

Ectopic lingual thyroid

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ABSTRACT

Thyroid ectopy is characterized by the presence of thyroid tissue outside its normal position resulting from a defect of the thyroid diverticulum migration from the base of the tongue until its final pre-tracheal position. One case is presented in a 12-year-old girl patient who consults for a failure to thrive estimated at less than three standard deviations (SD). Bone age was estimated at 8 years late compared to chronological age. The hormonal assessment showed hypothyroidism with negative thyroid antibodies. Cervical ultrasound was revealed thyroid parenchyma pre-dominantly left in place while sweeping the area under chin showed a nodular formation of the base of the tongue. Thyroid scan with technetium 99 m showed a selective uptake of radiotracer in sublingual position. Cervical computed tomography revealed a posterior median sublingual mass spontaneously hyperdense and enhancing sharply after injection of contrast. Treatment with thyroxine allowed obtaining euthyroidism. This case asks us to be careful before aetiological diagnosis of hypothyroidism in children, because although this is rare, the presence of a thyroid parenchyma up to the cervical ultrasound does not eliminate the presence of ectopic tissue.

Keywords: Dysphagia, hypothyroidism, lingual thyroid, thyroid scan

INTRODUCTION

The ectopic thyroid was described for the first time, in 1869, by Hickman in a newborn.^[1] This is a rare embryological anomaly characterized by the presence of thyroid tissue outside its normal position resulting from a defect of the thyroid diverticulum migration from the base of the tongue, place of appeared, until its final pre-tracheal position. The most common sites of ectopic thyroid are lingual, sublingual, thyroglossal, pre-tracheal, laryngotracheal and laterocervical^[2] and sometimes sub-mandibular, retroperitoneal and exceptionally at esophagus, mediastinum, heart, aorta, adrenal glands, pancreas, gall bladder and skin.^[3-7] The co-existence of a lingual thyroid tissue and an orthotopic thyroid is extremely rare.^[1] We report a case of this uncommon entity due to the coexistence of a non-functional orthotopic thyroid tissue with and partially functional lingual thyroid.

CASE REPORT

A 12-year-old female patient with no family history, addressed

to our department for investigation of a failure to thrive. In the personal history of this child, there is dysphagia to solids with high selective aspiration food lasting for 1 year (since 2001), which justified a consulting ear, nose and throat (ENT) in March 2002 and revealed a mass at the base of the tongue with a hormonal balance in favor of a biological hypothyroidism. No hormone replacement therapy was initiated and the patient admits that she never represented to consultation until February 2006, when she was sent to us for failure to thrive. On admission, she weighed 26.00 kg (-2SD), her height was 132 cm (-3SD), and pubertal development estimated at S1 P1 A1 (according to Tanner) without signs of thyroid dysfunction or dyspnea, dysphonia and dysphagia. The neck palpation was normal but examination of the back oral cavity showed a training round, sessile, smooth, red, and covered with some telangiectasia on the surface [Figure 1]. The hormonal assessment showed TSHus to 12 μ IU/ml (0.25-3.1) and FT4-5.11 pmol/l (10-20) in favor of a peripheral hypothyroidism, the dosage of thyroid antibodies (thyroperoxidase antibody and thyroglobulin antibody) was normal as well as the thyroglobulin. Bone age was estimated at 8 years late compared to chronological age. The neck ultrasonography revealed a hypoechoic sublingual formation and revealed the presence of a small orthotopic thyroid parenchyma predominating on the left. Thyroid scan with technetium 99m (Tc-99 m) showed a selective uptake of radiotracer in sublingual position [Figure 2].

A cervical computed tomography (CT) was performed and showed

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a posterior median sublingual mass spontaneously hyperdense and enhancing sharply after injection of contrast [Figure 3].

The diagnosis of hypothyroidism device in connection with a lingual thyroid is retained and the child received a hormone replacement therapy with thyroxine at a dose of 75 µg/d. After 5 months of treatment, the TSHus control was 1.75 µIU/ml signifying the biological euthyroidism.

DISCUSSION

The thyroid gland appears after the 3rd week of embryonic



Figure 1: Examination of the oral cavity back showing round, sessile, smooth, red and covered formation with some telangiectasia on the surface

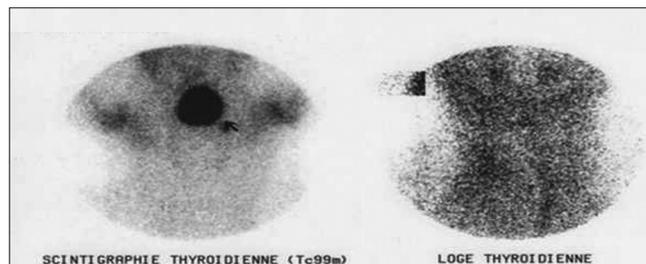


Figure 2: Thyroid scan with Tc 99m showing elective sublingual fixation of the radiotracer

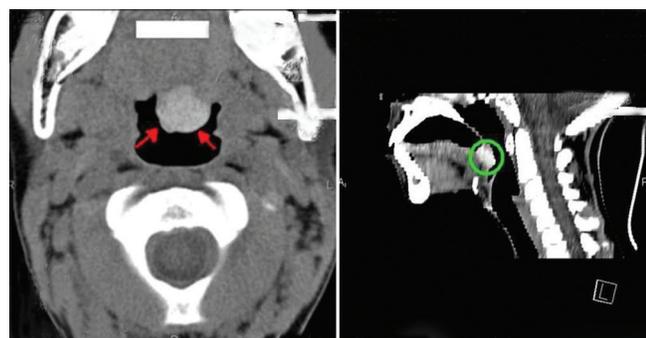


Figure 3: Posterior median sublingual mass spontaneously hyperdense and enhancing sharply after injection of contrast

development at the middle of the floor of the primitive pharynx (foramen cecum) and then migrates along the thyroglossal duct to reach its final anatomical position to the seventh week.^[8] Any disruption of this migration can result in ectopic thyroid. The lingual thyroid represents 90% of ectopic thyroid.^[9] In 70% of ectopic thyroid, the gland is not found in orthotopic position.^[1] The prevalence of lingual thyroid is 1/100000-300000 of the normal population, most frequently affect women, especially during adolescence and pregnancy,^[1] with a sex ratio female/male = 4/1.^[1,10] The preferred age of onset of clinical symptoms is 40 years with two statistical peaks at 12.5 and 50 years.^[11] Our patient is aged 12.

Patients with thyroid ectopy are usually asymptomatic, some cases are diagnosed incidentally.^[11] Two main clinical presentations can be seen, depending on the nature and the volume of functional ectopic thyroid tissue, hypothyroidism and pseudo-tumor. Furthermore, 70% of lingual thyroid are associated to hypothyroidism and 10% to cretinism.^[1] Our patient has hypothyroidism but without cretinism. On physical examination, the existence of a posterior lingual palpable mass evokes diagnosis, but is rarely noticed. A differential diagnosis is necessary with certain conditions such as thyroglossal duct cyst, lymphoma, fibroma, hemangioma, a tumor of the lingual salivary gland, a cancer of the tongue, and an epidermal cyst.^[8] Large volume ectopias can escape the new-born screening and be responsible for a considerable delay diagnostic.^[8]

In an older child, the attention may be drawn to unexplained short stature, as is the case of our patient, delayed puberty or psychomotor retardation.

Imaging technics most frequently used for the exploration of a lingual thyroid are thyroid scan with Tc-99m or iodine-123, CT and magnetic resonance imaging (MRI).

Thyroid scan with Tc-99m or iodine-123 or iodine-131 remains the best choice because it allows the topographic diagnosis of ectopic thyroid and confirm the presence or not of orthotopic thyroid gland. Furthermore, thyroid scan is important for differential diagnosis between ectopic thyroid tissue and other neck masses (as the fixation of the radiotracer at the base of the tongue witch confirms the nature of the ectopic thyroid tissue).^[7,11,12] In addition, for the radiotracer, Tc-99m is less radioactive than iodine-131 and allows a better visualization of images, has a lower cost, a shorter period of review, reported in children (as was the case in our patient) but its disadvantages are its accumulation in the salivary glands, making diagnosis difficult in the case of small masses. Iodine-123 is indicated in children but is not available in all centers (our case) and high costs limit its use.^[11]

CT and MRI are indicated for the pre-operative assessment when indicated surgical to localize the ectopic tissue and to evaluate the extension, they also keep their interest in case of intrathoracic goiter and in case of suspicion of a tumor process.^[7]

The cervical ultrasound enhances the sensitivity of the diagnosis of ectopic thyroid in the detection of hypervascularity.^[1]

The ectopic thyroid may be asymptomatic but the disease can also be studded with complications related to their location. In the case of a lingual thyroid, the patient may report feelings of foreign bodies, obstruction of the upper airway with dyspnea, dysphagia and food aspiration (the case of our patient), bleeding or ulceration that are secondary to the achievement of vessels on the surface of the gland.^[1] Malignant degeneration is exceptional with a similar risk to that of an orthotopic gland, only 33 cases have been reported in the literature with greater frequency for the vesicular type, Yaday reported the case of a 45-year-old woman, carrying a medullary carcinoma whose primary site was the lingual thyroid.^[1,13]

The pathogenesis of ectopic thyroid is still not clear. Some authors suggest that maternal immunoglobulins directed against thyroid antigens are the cause of this arrest of migration. Molecular abnormalities are also involved in these migration defects, the mutations in genes involved in thyroid development and differentiation are Nkx 2-1, Nkx 2-5, PAX8, Foxe1.^[14]

All these anomalies will lead to a partial or no migration of thyroid diverticulum. With regard to our child, the gland was ectopic in tongue position and appeared to result from a complete lack of migration. However, the presence of a thyroid parenchyma in the normal position is difficult to explain and may reflect a defect in the formation of organizational thyroid. Such an association is extremely rare since, this gland is the only ectopic thyroid tissue in 70% of cases.

For what concerns the therapeutic component, no consensus was reached^[7] in euthyroid patients who are asymptomatic and require simple monitoring for an enlarged gland or the appearance of possible complications.^[7,11] Medical treatment, based on a hormone replacement therapy with thyroxine, aims both to restore thyroid function and improve growth in stature^[7] and secondly to avoid development of ectopic tissue to prevent a surgical decision sometimes complex and will therefore, depend on the size of the gland and the presence of signs of compression.^[7,10] Total thyroidectomy is recommended by some authors because of risk of malignant degeneration.^[15] Iodine-131 is an alternative to surgery if it is against-indicated or impossible due to anatomical constraints or patient refusal. It is however to be avoided in children and its side effects are not insignificant.^[15]

Our child, in front of the disappearance of obstructive symptoms, the indication for surgery has not been raised and hormone

replacement therapy was the only treatment required with regular monitoring.

CONCLUSION

Our case report highlights a rare and unusual situation because of the coexistence of a non-functioning thyroid tissue and a partially functional lingual thyroid. The pathogenesis of such ectopias is still unclear. Ectopic lingual thyroid should be considered in any child's hypothyroidism and even in adults especially in the absence of thyroid antibodies.

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